

Screening for sensorineural deafness by health visitors

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Abstract

Screening for hearing loss in the first year of life, using the distraction test, remains the responsibility of health visitors in most health districts in the United Kingdom. We have evaluated the screening procedure used routinely in one health region in a population of infants at increased risk of sensorineural deafness. They were infants who weighed less than 2000 g at birth or infants who weighed 2000 g or more at birth and who spent more than 24 hours in a special care nursery. The infants' responses to a distraction test were recorded by health visitors and sent to the project office. The results were compared with information from a regional register of early childhood impairment that included children in whom sensorineural deafness had been diagnosed before the age of 3 years. The register had been compiled using information from a wide range of sources.

When used in this high risk population the distraction test was sensitive (91%), but non-specific (82%). The effectiveness of the screening programme was limited, however, because there was an increased risk of deafness among infants who missed being screened by health visitors. In addition, 71% of the deaf infants on the register were not in the high risk population.

Despite considerable advances in techniques for the screening of newborn infants for sensorineural deafness, screening for hearing loss still remains the responsibility of health visitors, who use the distraction test when the children reach the age of 6-8 months. There is evidence that this technique lacks sensitivity, and deaf children may be missed.^{1 2} The test also has a low specificity for sensorineural deafness, a large number of children having false positive results.^{3 4} Although some of these children will have a conductive hearing loss, it is expensive to retest or refer the infants who fail the test, and the parents become anxious.

Although the test is used by health visitors throughout the country,⁵ few health districts systematically collect information on test results and relate these to the hearing of the children.⁶ This means that a test of doubtful validity is being applied to thousands of infants each year with little attempt to evaluate either its accuracy in detecting deaf children or the effectiveness of programmes that include it.

We have studied the use of the distraction test in a population of infants considered to be at

increased risk of sensorineural deafness. By comparing test results against cases of sensorineural deafness identified from a regional register of impairment that was compiled concurrently, we have assessed the accuracy of the test, and have identified factors that limit the effectiveness of a community based hearing screening programme.

Subjects and methods

A subpopulation of infants considered to be at particular risk of sensorineural deafness were identified. They were infants born to mothers resident in the Oxford region in 1984 and 1985 who weighed less than 2000 g at birth, or who weighed 2000 g or more and spent longer than 24 hours in a special care nursery. Infants were identified by telephoning all 10 special care nurseries in the region once a week. Infants weighing less than 2000 g at birth who were not admitted to special care nurseries or who were born outside the region were identified from birth registration data. Information about infants weighing 2000 g or more who were admitted to special care nurseries outside the region was provided by health visitors.

Parents' consent for the transfer of information on the results of screening tests from the health visitor to the project office was sought at the time of discharge from the nursery; an 'opting out' approach was used.⁷

Health visitors were asked to apply the distraction test to this subpopulation using the technique routinely taught in their district. The birth history of each infant was known to the health visitor before testing.

We had noticed some variations in technique and the timing of tests, and these have been described previously.⁸ In two districts the test is routinely carried out at the age of 7 months, and in six districts at the age of 8 months. We asked health visitors to test at the age selected by their district and not to make any correction for gestational age at birth.

Results of the tests were recorded on a simple form and sent to the project office. Health visitors continued to screen the remaining infants in the population routinely, but the results of these screening tests were not available to us.

We asked health visitors to retest infants or refer infants for a specialist opinion whenever they considered it was necessary. If the infant did not turn to a sound made at one or both ears, this was regarded as a failure. In general, health visitors retested infants who had failed one test. Infants were usually referred after two

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test failures. Information about results of retests and intended referrals was also sent to the project office.

Concurrently a regional register of early childhood impairment, including sensorineural deafness, was set up. Cases were reported from several sources.⁹ At the age of 3 years we ascertained the hearing of each child, together with information on residence at the time of birth, birth weight, and admission to a special care nursery. The definition of sensorineural loss was a loss of 30 dB or more at any frequency in the range 0.5 to 4 kHz in one or both ears. In the absence of a pure tone audiogram, all children with a hearing aid fitted for sensorineural loss were included. It was possible that some deaf children were not on the register by the age of 3 years, particularly those with progressive hearing loss or high frequency loss that was detected later.

Those children on the register who had either confirmed congenital sensorineural deafness or early acquired sensorineural deafness, who were resident in the region at the time of birth, and were from the high risk population, were used as the 'gold standard' against which the screening test was assessed.

Results

STUDY POPULATION

Of the 64 881 infants born alive in 1984 and 1985 to mothers resident in the Oxford health region at the time of delivery, 4527 (7% of all live births) were eligible for enrolment into the study at the time of discharge from hospital. Of the eligible population, 1133 (25%) weighed less than 2000 g at birth. The remaining 3394 were larger infants who had been admitted to special care nurseries.

Forty eight infants died between the time of discharge from hospital and the age at which the distraction test was due (7 or 8 months). Of the remaining 4479 infants, 4116 had a distraction test. Reasons for failure to be tested are shown in table 1.

CHILDREN WITH SENSORINEURAL DEAFNESS

Sixty two children born during 1984 and 1985 were identified from the regional register as having sensorineural deafness at the age of 3 years. The risk of sensorineural deafness was higher among children who had weighed less than 2000 g at birth and also among larger infants who had required special care for longer than 24 hours (fig 1) than among the remainder of the infant population. Most of the deaf children (44/62), however, were not in these high risk groups.

Table 1 Number of infants eligible, available for testing, and tested

No (%) eligible for testing:		4527	(100)
Died before age of 8 months	48		
No (%) available for testing:		4479	(99)
Parents refused to cooperate	100		
Moved—not located	90		
Severe medical illness	11		
Not tested for unknown reason	162		
No (%) tested		4116	(91)

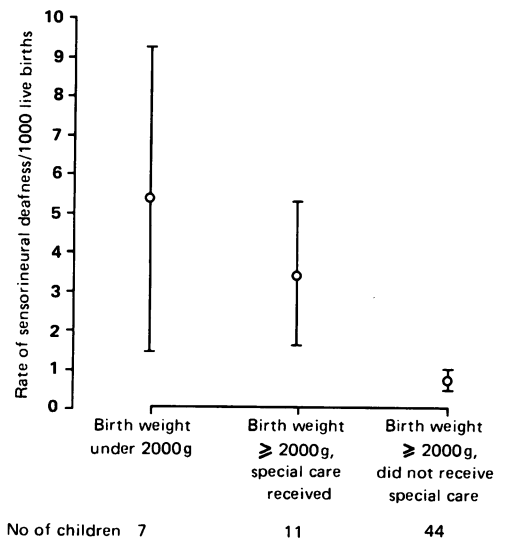


Figure 1 Incidence of sensorineural deafness, 1984-5. Values are expressed as mean, 95% confidence intervals.

There were 18 deaf children in the high risk subpopulation who were eligible for this screening programme. Seven of these children (39%) were among the 268 infants who had not been screened; none of the seven had been diagnosed as deaf or had hearing aids by the age of 8 months, although two had been seen in an audiology department.

EVALUATION OF DISTRACTION TEST IF APPLIED ONCE

Overall, 752 (18.3% of the screened population) failed the distraction test when it was first applied. There was no association between the age at screening and the proportion of infants who failed the screening test (fig 2), but the failure rate increased with decreasing gestational age (fig 3).

Of the 11 deaf infants in the population who were tested, 10 failed the distraction test and one passed when first tested. Characteristics of the distraction test when first applied are shown in table 2.

EVALUATION OF DISTRACTION TEST WITH AN OPTION TO RETEST

A total of 578 infants were retested. Of these, 525 had failed their first test, and of these 132 failed a second test. Fifty three of the 578 retested infants had passed the first distraction test and four of these failed on the second test (table 3).

All 11 deaf infants were retested. Ten of these failed the second distraction test and the same infant who had passed the first test passed the retest. The specificity of the distraction test improved with the retest option, however, although the positive predictive value remained low (7.6%).

EVALUATION OF THE POLICY OF APPLYING THE DISTRACTION TEST TO ALL INFANTS IN THE HIGH RISK POPULATION

Of the available population 363 (8%) were not

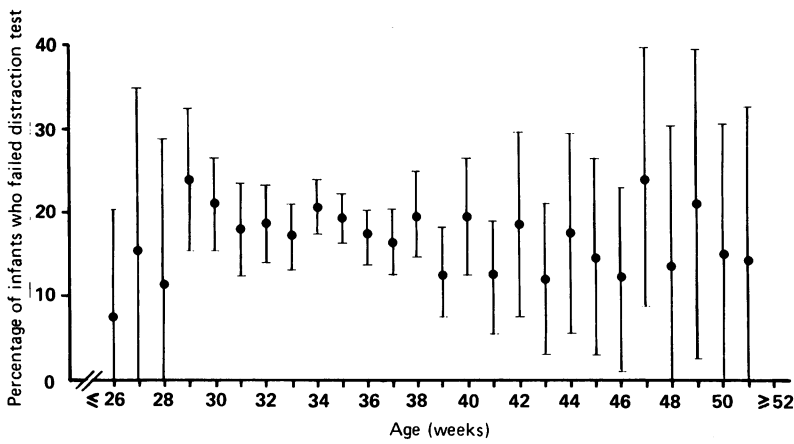


Figure 2 Proportion of infants who failed distraction test, and age tested. Values are expressed as mean, 95% confidence intervals.

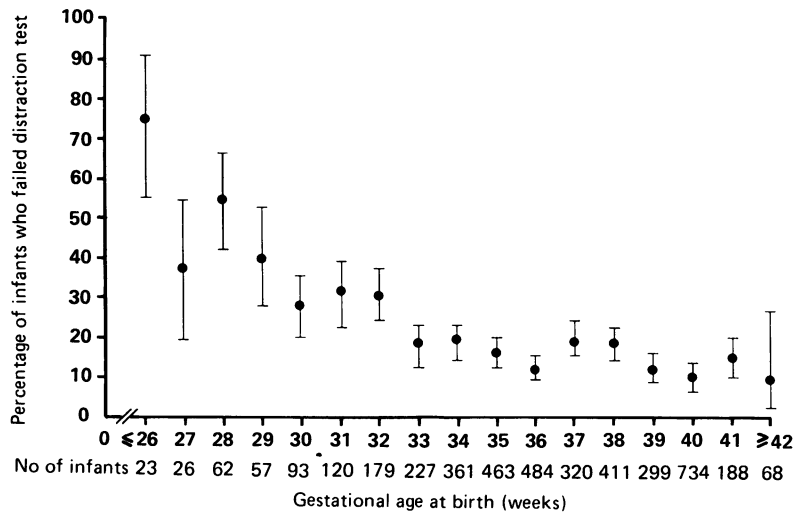


Figure 3 Proportion of infants in who failed distraction test, and gestational age at birth. Values are expressed as mean, 95% confidence intervals.

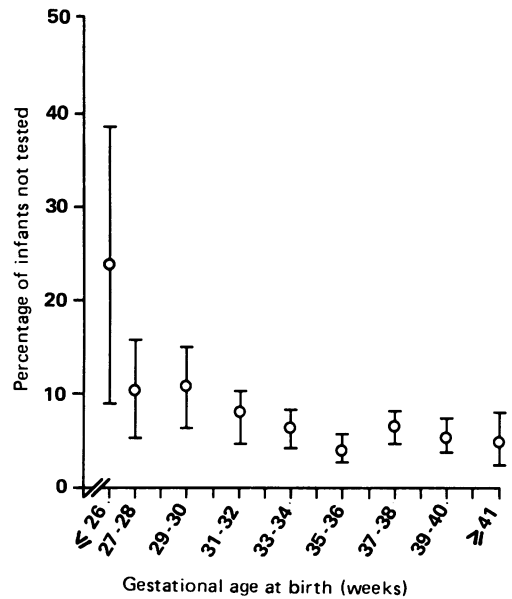


Figure 4 Proportion of infants who did not have distraction test, and gestational age at birth. Values are expressed as mean, 95% confidence intervals.

sensorineural deafness was considerably decreased (table 4). Sensitivity was reduced to 56%.

EVALUATION OF POLICY OF APPLYING THE DISTRACTION TEST TO ALL INFANTS IN HIGH RISK GROUP NOT ALREADY KNOWN TO AUDIOLOGY DEPARTMENTS

Two of the 11 deaf infants who were tested were already known to audiology departments although had not yet been diagnosed, and they both failed two distraction tests. On the premise that the purpose of screening is to detect pre-

Table 2 Distraction test at age 7-8 months: first time applied

	Sensorineural deafness		Total
	Present	Absent	
First distraction test:			
No who failed	10	742	752
No who passed	1	3363	3364
Total	11	4105	4116

Sensitivity 91%, specificity 82%, positive predictive value 1.3%, negative predictive value 99.9%.

Table 3 Distraction test with an option to retest

	Sensorineural deafness		Total
	Present	Absent	
No who failed both tests	10	122	132
No who passed one or both tests	1	3983	3984
Total	11	4105	4116

Sensitivity 91%, specificity 97%, positive predictive value 7.6%, negative predictive value 99.9%.

screened. The risk of not being screened increased with decreasing gestational age (fig 4). As seven of the 18 deaf infants in the population under study were not screened, the effectiveness of the policy to screen high risk infants for

Table 4 Screening policy applied to high risk population

	Sensorineural deafness		Total
	Present	Absent	
No who failed distraction test	10	122	132
No who passed distraction test	1	3983	3984
No not tested	7	356	363
Total	18	4461	4479

Sensitivity 56%, specificity 97%, positive predictive value 7.6%, negative predictive value 99.9%.

Table 5 Screening policy when applied to a high risk population, excluding infants already suspected of having an impairment

	Sensorineural deafness		Total
	Present	Absent	
No who failed distraction test (deafness not previously suspected)	8	122	130
No who failed distraction test (deafness already suspected)	2	0	2
No who passed distraction test	1	3983	3984
No not tested	7	356	363
Total	18	4461	4479

Sensitivity 44%, specificity 97%, positive predictive value 6.1%, negative predictive value 99.9%.

viously unsuspected impairment, exclusion of these two infants lowers the sensitivity still further (44%) (table 5).

ESTIMATE OF THE EFFECTIVENESS OF A POLICY OF SCREENING FOR SENSORINEURAL DEAFNESS IN THE TOTAL POPULATION AGED 8 MONTHS

If we assume that the distraction test will behave in the same way in the total population as it does in a high risk population, and that the proportion of children not tested or already known to audiology departments will be similar in the two populations, then only 27 of the 62 deaf children (44%) in the population will be detected by the policy of applying a distraction test. Further, as the prevalence of sensorineural deafness in the total population (62/64881:1.0/1000 live births) is lower than the prevalence in the high risk population (18/4854:3.7/1000 live births), the positive predictive value of the test when applied to the total population would be only 0.5%.

Discussion

It is generally agreed that early detection of sensorineural deafness is desirable because early provision of hearing aids seems to improve linguistic, intellectual, social, and emotional development.¹⁰ There has been concern about the lack of accuracy and reliability of methods currently used in community based hearing screening programmes as they may lead to late detection of sensorineural deafness.¹¹

There have been at least three responses to this concern. First, there have been considerable advances in the technique of neonatal hearing screening using an auditory response cradle,¹² auditory brain stem responses,¹³ and evoked otoacoustic emissions.¹⁴ If these techniques prove to be sensitive and specific and can be readily and inexpensively applied to the total newborn population, then the responsibility for early diagnosis could be shifted from the community to the newborn nursery. At present, however, these methods tend to be used only in selected special care nurseries as they are expensive to apply to the total population.¹⁵

An alternative response to the problem of late detection of deafness is to look for methods of improving the reliability of the distraction test. Training of health visitors and improved techniques in applying the test could reduce the proportion of infants who need referral for further assessment to 2.5% without loss of sensitivity of the test.¹⁶

A third strategy has been a take heed of parents' observations and concerns by asking them directly about their suspicions of lack of hearing,¹⁷ by developing check lists for use in the early months of life,¹⁸ and by encouraging parents to take children directly to specialist clinics.¹⁹

At the time of this study the policy of applying a distraction test to detect sensorineural deafness was widely accepted throughout the region. Although the test when applied to a high risk population seemed to have a high sensitivity (91%), there were factors that severely

limited the effectiveness of such a screening programme.

First, almost a fifth of the infants tested failed the test. By retesting the failure rate could be reduced to 8% of the population tested. This lack of specificity is expensive as the infants have to be recalled for second testing or referred to specialist clinics. Another aspect of the high failure rate is that a failed test can cause anxiety and concern among parents. There may also be disbelief in the test result and doubt in the minds of parents as to the value of a test that so many infants fail. In a study of parents' views of infant screening, we frequently found this type of response—'He failed the test, but I know he can hear' (F Sherratt, personal communication). This attitude could lead to low compliance in attending for further testing.

A second factor that limits the effectiveness of the screening programme is the number of hearing problems among infants who were not screened. Over a third of deaf infants were among the 8% of the high risk population who were not screened.

The risk of not being screened increased with decreasing gestational age at birth. There may be a number of reasons for this. Infants of low gestational age at birth tend to have delayed motor development during the first year of life. The distraction test requires that the infant be sitting and have sufficient neck and trunk control to turn to the sounds. We have shown that health visitors tend to delay applying the tests in preterm infants, presumably until they judge that this developmental level to have been reached.²⁰ It is likely that once infants are out of the routine pattern of testing at 7 or 8 months of age, there is an increased risk of their not being tested. Some infants remain in hospital, sometimes on a ventilator, beyond the age of routine testing, and special provision needs to be made for them. A third reason for not testing these high risk infants within the community is the belief that infants who are attending follow up clinics will have their hearing checked by the hospital based specialist services. Although this may be a routine in some hospitals, it is by no means a universal practice. Better communication between hospital specialist and follow up services and the community services might help to prevent the infant not being tested by either service.

A further important factor in the failure to screen infants is population migration: two of the deaf children who failed to be tested had changed their address and had not been relocated by the age of 8 months. As cross boundary exchanges of records tends to be slow, it may be that parent held records, together with a better appreciation by parents of the importance of testing, could be useful.

Overall it would seem that infants who miss being screened for whatever reason need to be identified, located, and tested as they have a higher than average prevalence of impairment.^{21 22}

A third factor that can limit the effectiveness of a screening programme is inappropriate 'targetting'. Although low birthweight infants and infants who have had adverse perinatal

factors are at increased risk of sensorineural deafness,^{23 24} most deaf infants will not have been admitted to a special care nursery. This has important implications for screening whether in the newborn period or later in infancy. Our screening programme missed 70% of the deaf children on the regional register.

It is possible that the performance of the distraction test might differ in the high risk population compared with the remainder of the infant population. A higher failure rate in the high risk population might be expected for two reasons: immature infants may be unable to give the motor response to the sound stimulus that is necessary for a test pass and health visitors were aware of the early history of the infant and of the increased risk of deafness in the high risk group. This could have altered the threshold of suspicion resulting in a higher true positive and false positive rate, and therefore a higher sensitivity but lower specificity, in the high risk group than in the rest of the population.

Finally, some deaf infants who failed the distraction test and had been appropriately referred to their general practitioners were undiagnosed by 18 months of age and were often not reported as a 'case' to the regional register until after their second birthdays. One of the criteria for a screening test is the provision of diagnostic services and treatment for those who fail the test. Without this back up support a screening programme will fail, however accurate the test itself.

We conclude that the distraction test as routinely applied by health visitors can be a sensitive if somewhat non-specific screening test. The effectiveness of a screening programme using the test is severely limited however, by failure to test all the infant population. Screening that is aimed only at infants who needed special care at birth will miss a high proportion of the deaf children in the community, and high risk infants who miss being tested seem to be particularly vulnerable.

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